Transarterial Wedged-catheter, Flow-arrest, N-butyl Cyanoacrylate Embolization of Three Dural Arteriovenous Fistulae in a Single Patient

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Summary

The pathogenesis of dural arteriovenous fistulas (DAVFs) is currently unknown, with multiple DAVFs being rare. For patients with limited venous access secondary to sinus thrombosis, or for patients where parent sinus occlusion would not be tolerated, transvenous embolization may not be possible and other treatment methods must be considered. A 69-year-old female patient with a two-year history of progressive headaches, memory loss, and unsteady gait underwent cerebral angiography that revealed three separate DAVFs with congested cortical venous drainage overlying both frontal lobes. Using an application of a transarterial wedged-catheter, flow-arrest technique, N-butyl cyanoacrylate was deposited across all three pathologic arteriovenous connections providing a definitive cure. Transarterial NBCA embolization may provide curative treatment of DAVFs, and is of particular utility in situations where access to the draining venous structures is limited.

Introduction

Dural arteriovenous fistulas (DAVFs) are acquired lesions that can cause neurologic deficit and intracranial haemorrhage [1]. The pathogenesis of DAVFs is uncertain, but, venous sinus thrombosis, venous hypertension, haemodynamic forces, congenital dysplasia, angiogenic growth factors, trauma, and cranial surgery are factors in their pathophysiology and natural history [2-14]. Although recent studies have reported the incidence of multiple DAVFs [7,15-17], there is limited information regarding the radiographic characteristics and treatment options for patients with these complex lesions. Furthermore, transvenous endovascular [18,19] and open surgical techniques [20-22] for the treatment of DAVFs have been well described, however, new transarterial techniques aimed at curing DAVFs have not received much attention in the literature [23]. Therefore, we also discuss an application of a transarterial wedged-catheter, flow-arrest, N-butyl cyanoacrylate (NBCA) embolization technique [24], which was used as curative treatment for all three DAVFs.

Clinical Presentation

History

The patient is a 69-year-old female with a past medical history of well-controlled hypertension and hypothyroidism who presented with a two-year history of progressive bifrontal headaches, memory loss, and unsteady gait. Her neurologic examination was significant for a wide-based gait and unsteadiness when walking heel-to-toe. An MRI revealed multiple flow voids on the medial surface of the left frontal

lobe consistent with a vascular malformation. There was no evidence of T2 signal abnormality within the brain parenchyma. She was referred to our institution for angiographic evaluation and treatment.

Angiographic Evaluation

Cerebral angiography demonstrated three independent DAVFs localized to: 1) the anterior falx, 2) the parasagittal right frontal convexity and 3) the middle right frontal convexity (figure 1 and table 1).

The anterior falcial shunt (Cognard grade IV)[1] was supplied through the anterior falcial artery, which was vascularized primarily through ethmoidal branches of the ophthalmic and sphenopalatine arteries bilaterally, the left middle meningeal artery, and transosseous branches of both superficial temporal arteries. Venous outflow resulted in extensive venous congestion over the left frontal lobe draining through ectatic cortical veins into the deep and superficial left Sylvian veins, superior sagittal sinus, and through the vein of Labbe into the left transverse sinus.

The right parasagittal DAVF (Cognard grade III) was supplied by an anterior frontal branch of the right middle meningeal artery, and emptied through non-ectatic cortical veins into the superior sagittal sinus. The third, middle right frontal convexity DAVF (Cognard grade III) was supplied by a posterior frontal branch of the right middle meningeal artery and drained through a dural vein into the right superficial Sylvian veins.

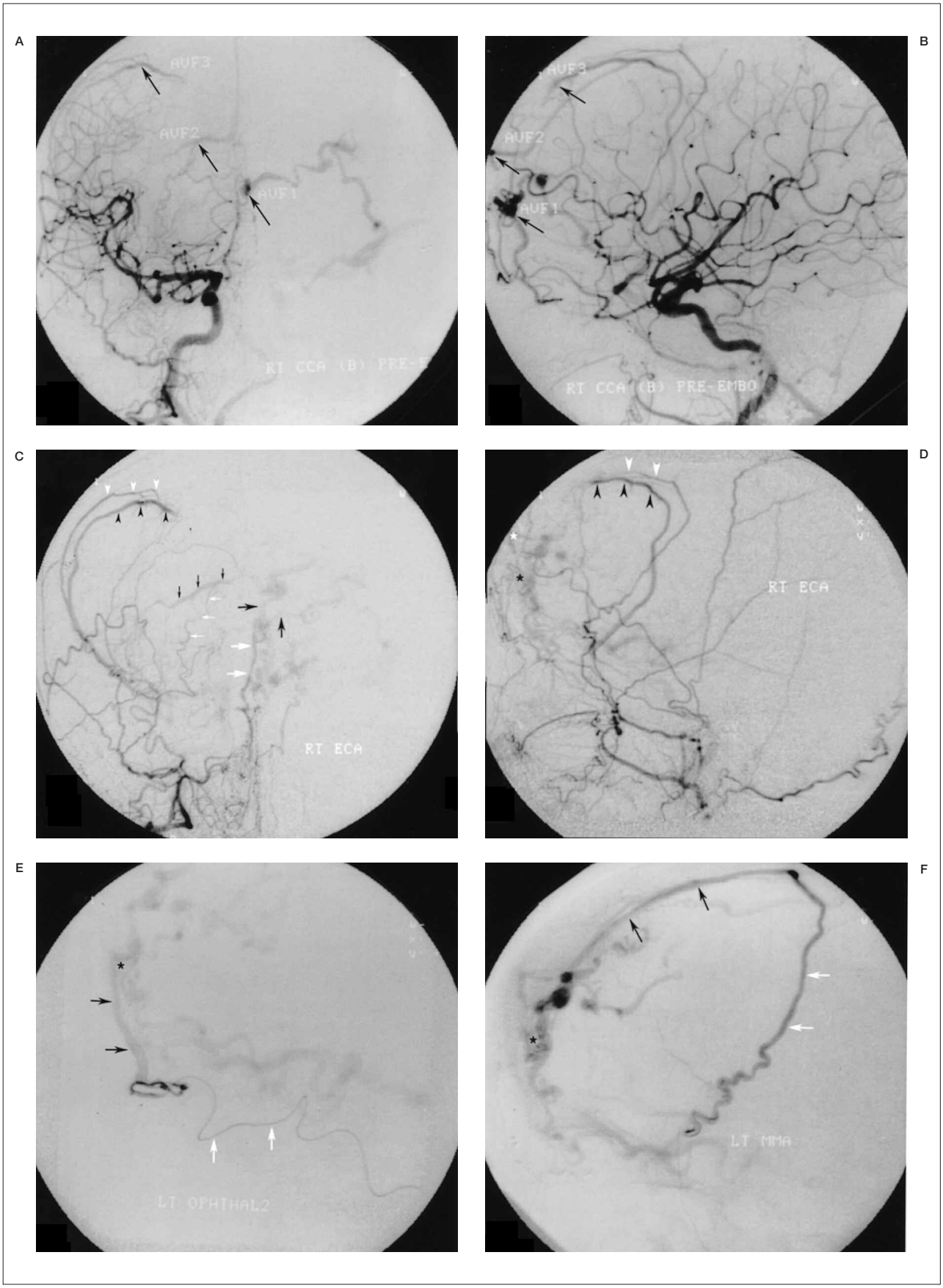
The patient's headache was likely caused by abnormal vascular engorgement of her falx and dura. The etiology of her memory loss and un-

Figure 1 Pre-embolization right common carotid injection, frontal (A) and lateral (B) projections, demonstrating three separate DAVFs with direct cortical venous drainage. The location of each fistula is indicated (black arrows). A selective right external carotid injection, frontal (C) and lateral (D) projections, characterizes the arterial feeders and venous drainage of each fistula. In the frontal projection (C), DAVF #1 (anterior falcial) is fed by the right anterior falcial artery (large white arrows) and drains into left hemisphere cortical veins (large black arrows). DAVF #2 (right parasagittal) is fed by a branch of the right middle meningeal artery (small white arrows) and drains into a right hemisphere cortical vein (small black arrows). DAVF #3 (right middle frontal) is fed by a branch of the right middle meningeal artery (white arrowheads) and drains into a right hemisphere cortical vein (black arrowheads) and then into a right Sylvian vein. In the lateral projection (D), the arterial feeder (white arrowheads) and venous drainage (black arrowheads) of DAVF #3 is evident. The location of DAVF #1 (white star) and DAVF #2 (black star) is also illustrated. (E) Lateral projection, selective left ophthalmic artery injection demonstrates arterial supply to DAVF #1 via the anterior falcial artery (black arrows). The pathological arteriovenous connection is evident (black star), as is the microcatheter in the ophthalmic artery (white arrows). (F) Lateral projection, selective left middle meningeal injection, demonstrates additional arterial supply (black arrows) to DAVF #1 from the left middle meningeal artery (white arrows).

Table 1 **Characteristics and endovascular treatment of three DAVFs in one patient**

Location	Grade [1]	Arterial Supply	Venous Drainage [2]	Venous Ectasia	Primary NBCA Injection [3]	Prepatory Embolizations [4]
DAVF #1 anterior falcial, interhemispheric	IV	B/L anterior falcial B/L sphenopalatine B/L superficial temporal L middle meningeal	L cortical veins L cavernous sinus L superior sagital sinus L transverse sinus	present	ethmoidal division of R ophthalmic	ethmoidal branches of R sphenopalatine - PVA L middle meningeal - NBCA ethmoidal division of L ophthalmic - NBCA
DAVF #2 R parasagittal frontal convexity	III	R middle meningea	R cortical veins superior sagital sinus	none	frontal division of R middle meningeal	none
DAVF #3 R middle frontal convexity	III	R middle meningeal	R cortical veins R superficial sylvian system	none	frontal division of R middle meningeal	none

Abbreviations: N-butyl cyanoacrylic (NBCA), bilateral (B/L), right (R), left (L), dural arteriovenous fistula (DAVF).
[1] Cognard C, Gobin Y, Pierot L: Neurological symptoms of intracranial dural arteriovenous fistulas: clinical and angiographic correlation in 205 cases. A revisited classification of venous drainage. Radiology, 1995, 194:671-680.
[2] The the primary venous drainage is listed in large font, followed by subsequent drainage pathways in small font.
[3] This column lists the arterial feeder that received the definitive wedged-catheter (flow-arrest) NBCA injection.
[4] This column lists any arterial feeders that were embolised prior to the definitive NBCA injection. The embolised vessel and type of embolic material are listed.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steady gait is less certain, however, venous hypertension was likely a contributing factor to these symptoms.

Intervention

After considering her progressive symptomatology and extensive cortical venous drainage, curative treatment of all three DAVFs was recommended. Due to the bilateral contributions from the ophthalmic arteries, embolization was staged to avoid potential loss of vision in both eyes.

During the first session, contributions from the right sphenopalatine, left ophthalmic, and left middle meningeal arteries were embolized with a combination of polyvinyl alcohol (PVA, Cordis Neurovascular, Miami, FL) and NBCA (Histoacryl, Yocan Medical, Toronto CA), significantly reducing collateral inflow into the anterior falcial shunt, however, not affecting curative occlusion. A wedged-catheter, flow-arrest NBCA injection was performed from an ethmoidal branch of the left ophthalmic artery, yet failed to transverse the pathological fistula secondary to collateral inflow causing glue fragmentation. The patient was then awakened and discharged from the hospital after overnight observation. Her subsequent ophthalmologic examination remained unchanged and she returned for definitive treatment four weeks later.

Angiographic evaluation at the start of the second session disclosed residual supply to the anterior falcial DAVF primarily through ethmoidal branches of the right ophthalmic artery with minor collateral contribution through transosseous branches of the superficial temporal artery. Definitive embolization of the anterior falcial lesion was performed from a right ophthalmic approach (figure 2A,B) with liquid embolic agent (NBCA) delivered through branches of the ethmoidal division supplying the remaining anterior falcial shunt. The curative embolization was facilitated by wedged-catheter, flow-arrest conditions, which permitted controlled hydraulic delivery of the NBCA through the targeted DAVF and into the immediate recipient vein, without reflux into the more proximal segments of the right ophthalmic artery or excessive distal venous embolization over the left frontal cortex. The right middle frontal convexity and parasagittal DAVFs were then occluded by single deposition NBCA embolization of the respective middle meningeal branches supplying each fistula under similar wedged-catheter, flow-arrest conditions. Guidewire directed microcatheters were used including the Prowler-10, Prowler-14, and Rapid Transit (Cordis Neurovascular, Miami Lakes, FL). The percent volume of NBCA in ethiodal was approximately 30-35% for all injections.

For all three depositions performed during this session the column of NBCA traversed the shunting site, thus occluding the fistula and proximal draining vein. Postembolization control angiography confirmed the occlusion of all three DAVFs (figure 2C-F). The patient experienced no complications and was discharged on postembolization day two. At one-year postembolization follow-up, the patient was neurologically intact and had no complaints.

Discussion

Multiple DAVF Pathogenesis

Although the pathogenesis of DAVFs is currently unknown, they appear to be acquired lesions with an ill-defined relationship to venous drainage abnormalities and specifically venous thrombosis [9,25,26]. Animal studies support the hypothesis that venous thrombosis may initiate DAVF formation [8,12]. According to this theory, venous hypertension and/or thrombosed sinus wall inflammation may up-regulate angiogenic growth-factor release, causing neovascularization of the affected sinus wall and increase thrombus formation. During recanalization of the previously thrombosed sinus, establishment

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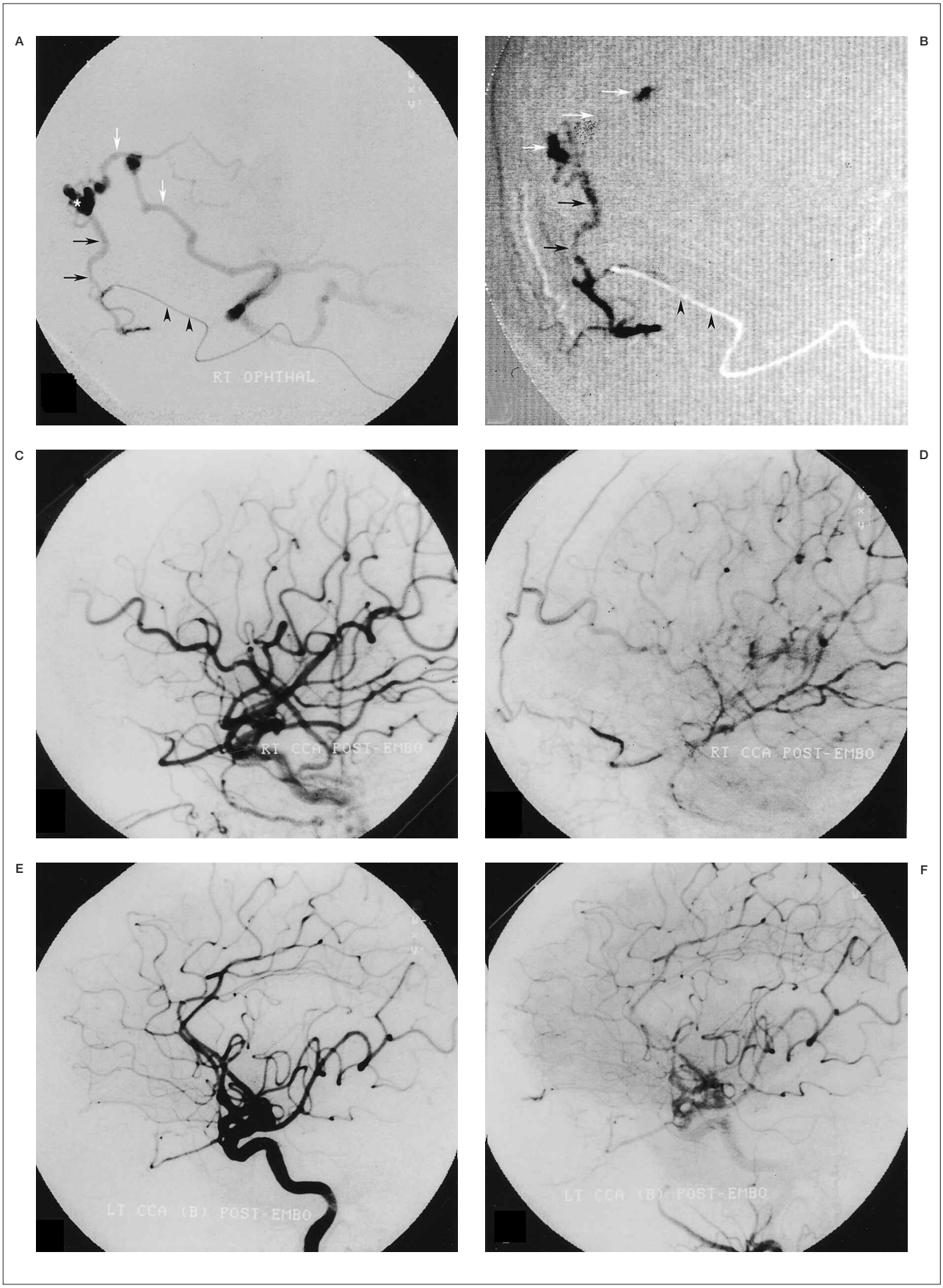
Figure 2 (A) Microinjection with the catheter wedged in an ethmoidal branch of the right ophthalmic artery, lateral projection, just prior to NBCA injection. The following structures are demonstrated: microcatheter within an ethmoidal artery (black arrowheads), anterior falcial artery (black arrows), pathological arteriovenous connection of DAVF #1 (white star), and cortical venous drainage (white arrows). (B) NBCA glue cast from the same microcatheter wedged in an ethmoidal division of the right ophthalmic artery, lateral projection radiograph. NBCA fills the ethmoidal branch distal to the wedged catheter tip, the anterior falcial artery (black arrows) and the proximal portion of the cortical draining vein (white arrows). The microcatheter track is also visible (black arrowheads). (C-F) Post-embolization control angiography following obliteration of all three DAVFs using flow-arrest NBCA injections from wedged-catheter positions. Right common carotid injection, lateral projection, early (C) and late (D) arterial phases. Left common carotid injection, lateral projection, early (C) and late (D) arterial phases. No evidence of arteriovenous shunting is present.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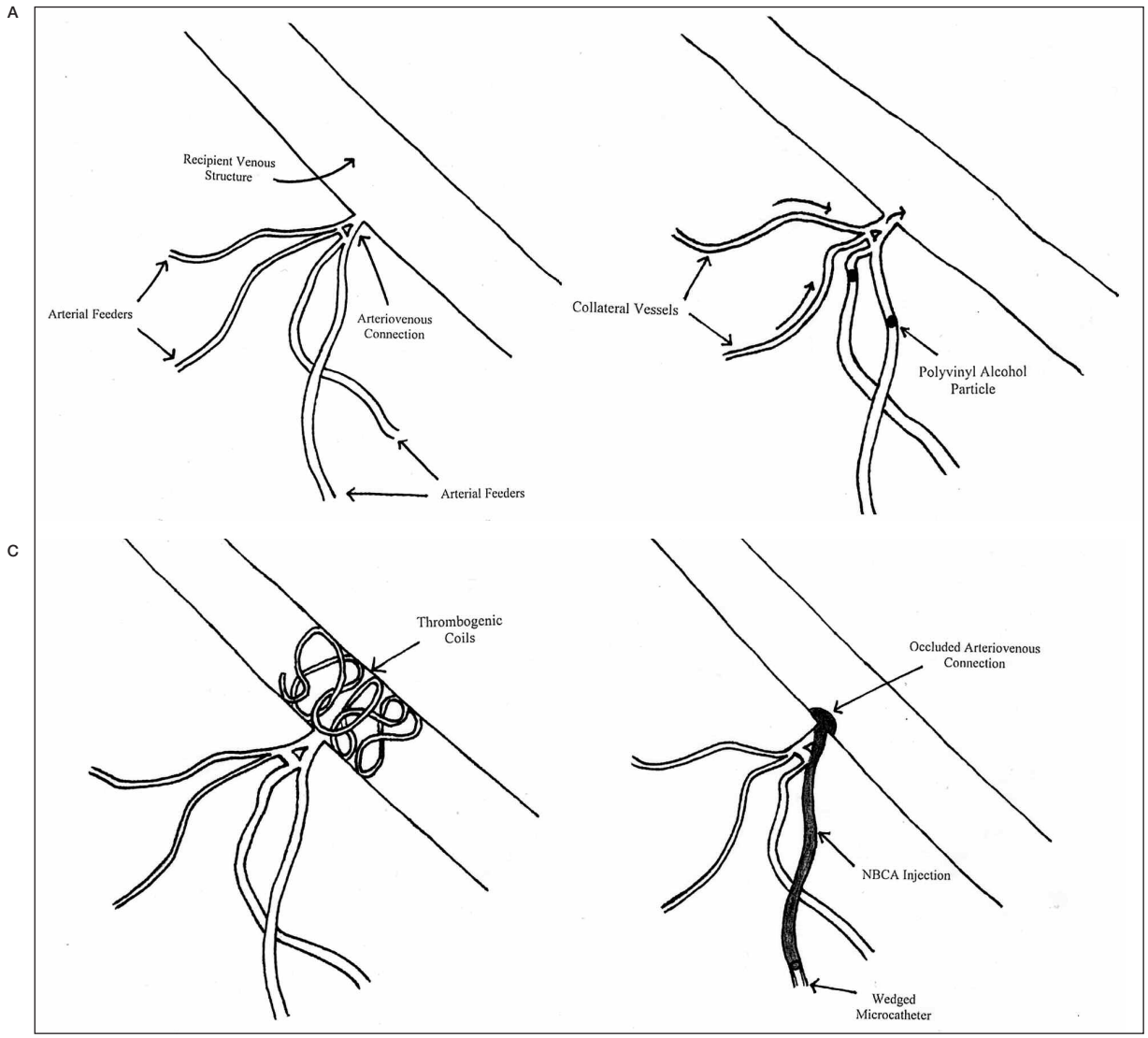

Figure 3 DAVF embolization techniques. (A) Schematic of a DAVF. (B) Proximal feeding artery occlusion with particulate embolization (no cure). (B) Transvenous coil embolisation of the parent venous structure (cure). (D) Transarterial wedged-catheter NBCA injection under flow-arrest conditions across the fistula entering the parent venous structure (cure).

cur [2,3,10]. A rat model of venous hypertension supports this theory, but it is uncertain whether these models reflect actual DAVF pathogenesis, or if they are experimental phenomena not causally related to the formation of the DAVF [8,12]. Conversely, DAVFs may induce recipient venous sinus occlusion secondary to abnormal haemodynamic forces causing intimal hyperplasia, thrombosis, and stenosis [15,27]. Therefore, venous sinus thrombosis may not be a causative event in DAVF formation, but a pathological response to an already established fistula.

Certain pathological entities, including DAVFs, diabetic retinopathy, arthritis, psoriasis, and haemangiomatosis, where strong angiogenic stimuli overcome homeostatic barriers causing unabated vascular proliferation may have a common pathogenesis and be categorized as "angiogenic diseases" [5,6]. In this pathologic state, excessive vascular growth can cause tissue destruction and profound symptoms. A case of pulmonary haemangiomatosis, a rare angiogenic disorder of progressive pulmonary haemangiomas that is usually fatal, was successfully treated with interferon, a potent anti-angiogenic drug that inhibits endothelial cell

proliferation and motility [28,29]. As an "angiogenic disease", DAVFs may also potentially be treated with an anti-angiogenic drug [3,6,7]. For DAVFs, specific anti-angiogenic therapy may promote spontaneous obliteration, hasten occlusion following stereotactic radiation, and prevent recurrence following successful treatment [3,6].

If DAVF formation reflects a general state of hypercoagulability, or vascular proliferative state, it could explain why our patient harbored three separate DAVFs. However, patients with multiple DAVFs are rare and only a few cases have been reported. Further research will hopefully be able to explain the pathogenesis of these complicated lesions.

DAVF Treatment

The indication for curative treatment of DAVFs is predicated by angiographically determined risk factors. Specifically, when DAVF outflow involves leptomeningeal veins, either directly or indirectly via reflux from a dural sinus, the risk of intracranial haemorrhage and venous hypertension induced neurological deficit or seizures substantially increases. In contrast, patients without cortical venous involvement usually follow a benign clinical course. Natural history studies have been performed that confirm these findings [30,31], and therefore, many patients without cortical venous drainage can be managed successfully with observation and periodic radiographic re-evaluation.

In order to cure a DAVF, its pathologic arteriovenous connection must be eliminated (figure 3). Proximal feeding artery ligation alone, via endovascular or surgical methods, is insufficient as the fistula recruits collateral sources of arterial supply. Transvenous embolization has become a proven treatment modality for DAVFs, obliterating the arteriovenous fistula by sacrificing the recipient venous structure [18,19,32]. For patients with limited venous access secondary to sinus thrombosis, or for patients where parent sinus occlusion would not be tolerated, transvenous embolization may not be possible and other treatment methods must be considered.

Open surgical management remains an efficacious treatment option for some DAVFs [20-22]. Determined by angiographic characteristics, two operative techniques may be used: 1) en bloc DAVF and parent sinus resection for sinusal DAVFs [22] or 2) selective arteriovenous disconnection for cortical DAVFs with direct leptomeningeal venous drainage [20,21]. Unfortunately, profound blood loss and postoperative recurrence remain problematic for surgically resected sinusal DAVFs; while for non-sinusal DAVFs, precise intraoperative localization and multiplicity of lesions may be prohibitive.

Our patient was treated with an application of a transarterial wedged-catheter flow-arrest embolization technique in which NBCA was injected, via an arterial feeder in a flow-controlled manner, across the fistula into the parent venous structure, thus eliminating the pathologic arteriovenous connection. As has been described for brain arteriovenous malformations [24], with this technique the arterial pedicle beyond the microcatheter tip becomes an extension of the catheter and allows for improved delivery of the liquid embolic agent across the fistula site. Recanalization should not occur if the liquid embolic agent completely occludes the fistula by crossing into the immediate receptive venous structure. The volume of NBCA entering the parent venous structure is minimized to prevent distal embolism. The permeation of the fistula with a liquid polymeric agent is preferable for the following reasons: 1) the fistula site is certainly occluded, thus reducing the likelihood of alternative venous drainage pathways being recruited, 2) is not limited by venous access problems, including thrombosed or stenotic dural sinuses, and 3) does not require the sacrifice of functional venous pathways that may drain normal brain parenchyma.

Reducing the degree of arteriovenous shunting prior to the definitive wedged-catheter (flow-arrest) injection minimizes the risk of uncontrolled distal venous embolization of NBCA. To minimize this risk, high-flow DAVFs may require prepatory embolization with PVA to decrease collateral contribution to the arteriovenous shunt prior to the definitive NBCA injection; thereby diminishing fragmentation of the glue column due to collateral inflow into the main arterial conduit during subsequent NBCA embolizations.

Conclusions

Transarterial wedged-catheter flow-arrest NBCA embolization is a potentially safe and effective treatment for intracranial DAVFs. It is of particular utility in situations where access to the draining venous structures is limited.

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